Arch Jpn Chir 58(2), 245～249, März, 1989

症　　例

Epidermoid Tumor Presenting with Trigeminal Neuralgia and Ipsilateral Hemifacial Spasm: A Case Report

Shin-ichi Otsuka, Shoji Nakatsu, Shigeo Matsumoto, Shin-ichi Sato, Takahiko Motozaki, Sadahiko Ban, Toyoshiro Yamamoto and Hirofumi Shirane*

Department of Neurosurgery,
*Department of Clinical Pathology, Kobe City General Hospital
Received for Publication, Jan. 10, 1989.

Abstract

A case of epidermoid tumor presenting with a painful tic convulsif was reported. A 35-year old male with trigeminal neuralgia and ipsilateral hemifacial spasm was diagnosed as having an epidermoid by CT and metrizamide CT cisternography and the symptoms were completely eliminated after the operation. In this case, metrizamide CT cisternogrphy was very useful for preoperative diagnosis by demonstrating the characteristic findings of the epidermoid. It should be taken into consideration that there are some cases with trigeminal neuralgia and/or hemifacial spasm whose symptoms are due to brain tumors.

Introduction

The term 'painful tic convulsif' was coined in 1920 by Cushing[2] to describe the coexistence of trigeminal neuralgia and ipsilateral hemifacial spasm. Only a few cases with painful tic convulsif caused by brain tumors have been reported. We have experienced a case with painful tic convulsif caused by epidermoid and in this case metrizamide CT cisternography was very useful for preoperative diagnosis. We described this rare case and review the relevant literature.

Case report

A 35-year-old male had occasionally felt mild pain in the right mandibular region for about 15 years. The pain had become more severe since around June, 1986, whenever he chewed or brushed his teeth. Under the diagnosis of trigeminal neuralgia, he was treated with administ-

Key words: Epidermoid, Painful tic convulsif, Metrizamide CT cisternography.
索引語：類上皮腫，疼痛性顔面痙攣，メトリザマイド CT 脳槽造影.
Present address: Department of Neurosurgery, Kobe City General Hospital, 4-6 Nakamachi, Minatojima, Chuo-ku, Kobe 650.

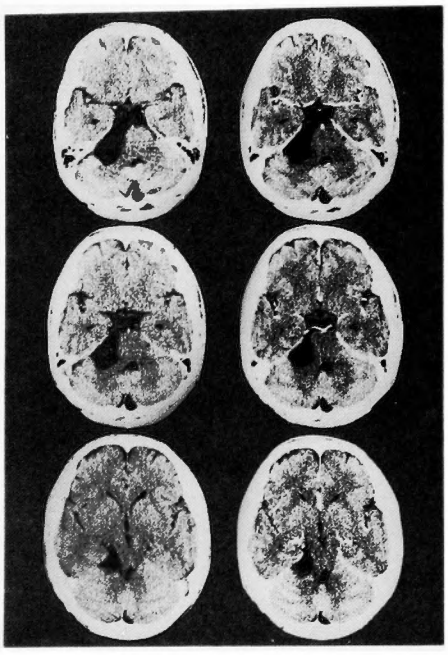

Fig. 1

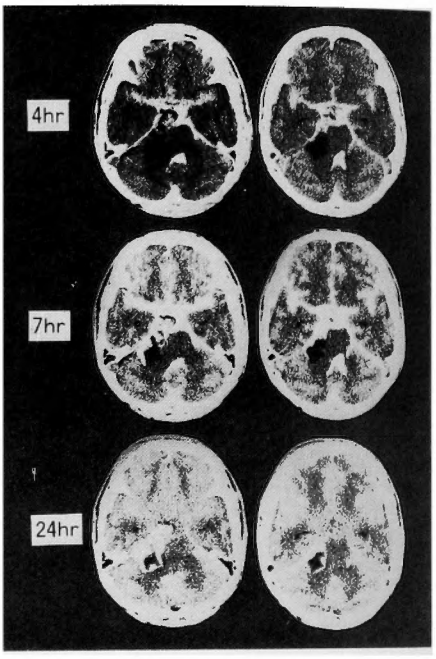

Fig. 2

Fig. 1 The plain CT (left side) and the enhanced CT (right side) revealed a low density mass in the right cerebello-pontine angle extending to the prepontine and the ambient cisterns. This low density mass was not enhanced after the intravenous injection of contrast medium.

Fig. 2 Metrizamide CT cisternography: Irregular spreading of contrast medium into the low density mass could be seen as time progressed after intrathecal injection.

ration of carbamazepine in December, 1986 and the pain was somewhat relieved thereafter. As he developed right hemifacial spasm since March, 1987 and brain tumor was identified by CT, he was admitted to our hospital on April 30, 1987.

The neurological findings on his admission revealed mild hypesthesia over the distribution of the third branch of the right trigeminal nerve and hemifacial spasm on the right side mainly around the right eyelid.

Skull X-rays were normal. CT revealed a low density mass in the right cerebello-pontine angle extending to the prepontine and the ambient cisterns (Fig. 1). This low density mass was not enhanced after the intravenous injection of contrast medium. Metrizamide CT cisternography showed irregular spreading of the contrast medium in the low density mass as time progressed after intrathecal injection (Fig. 2). The vertebral angiography showed the displacement of the vessels due to the mass effect of the tumor and there was no tumor stain.

On the basis of the above-mentioned results of the neuro-radiological examinations an epidermoid tumor was suspected and the extirpation of the tumor was conducted with right suboccipital craniectomy. A tumor with a typical pearly color was identified in the cerebellopontine angle. The V, VII and VIII cranial nerves were surrounded by the tumor. The tumor was removed piece-by-piece and these cranial nerves were completely decompressed.

Fig. 3 The histopathological findings revealed the structure composed of a stratified squamous epithelium and keratin debris.

The histopathological findings revealed the epidermoid tumor which was composed of stratified squamous epithelium and keratin debris (Fig. 3).

Postoperative course was favorable and both trigeminal neuralgia and hemifacial spasm were completely eliminated.

Discussion

CUSHING reported the cases with the coexistence of trigeminal neuralgia and ipsilateral hemifacial spasm as 'painful tic convulsif' in 1920[2]. Some other cases have been reported

Table 1 Reported cases of an epidermoid presenting with painful tic convulsif.

	Reference	Case (age, sex)	Side	Diagnostic Methods	Operation
1	Revilla (1947)	51 , F	Rt	not described	+
2	Gardner (1968)	54 , F	Lt	Skull X-P	+
3	Wakabayashi (1983)	49 , M	Lt	CT Metrizamide CT cisternography Angiography	+

since then although they are limited in number[1,3,5,6,7,9]. Our review of the literature showed that only 3 cases with painful tic convulsif due to an epidermoid have been reported[3,7,9] (Table 1). In 1947, REVILLA[7] reported a case of a 51-year-old female, while GARDNER[3] a case of a 54-year-old female in 1968 and in both cases diagnosis was confirmed by operations. WAKABAYSHI, et al.[9] reported a case of a 49-year old male. In this case, CT, metrizamide CT cisternography and angiography were performed before operation, and the tumor was completely removed by suboccipital craniectomy. The cases with painful tic convulsif caused by brain tumors other than epidermoid were reported by MAURICE-WILLIAMS[5], who described a case with angioma and one with acoustic neurinoma, but both of them only underwent neuroradiological examinations and none of them underwent operations, so the diagnosis was not confirmed. COOK, et al.[1] reported 11 cases with painful tic convulsif. Of these, 10 cases were not due to a tumor, while one case with meningioma was identified as being compressed by meningioma upon AICA, which then compressed the VII cranial nerve. Thus, the existence of some kinds of brain tumors was reported as the cause of painful tic convulsif, but the diagnosis was confirmed only in the cases with an epidermoid.

With regard to the findings of metrizamide CT cisternography on epidermoid, some reports have shown that the irregular high density areas were observed in the low density mass after intrathecal injection of contrast medium[4,8]. These findings seemed to indicate that the water soluble contrast medium entered the tumor through the tumor capsule or the defects of it and subsequently spreaded the depths of the tumor. SEKIHARA, et al.[8] reported a case with an epidermoid of the fourth ventricle, and in this case, infiltration of the contrast medium into the tumor was confirmed by measuring the concentration of metrizamide in the cyst fluid which was taken during the operation after the injection metrizamide into the lateral ventricle through the shunt valve just before the operation. These findings of metrizamide CT cisternography have never been reported for any other brain tumors and seemed to be important in differential diagnosis because these findings are characteristic for an epidermoid.

The present case revealed characteristic findings of metrizamide CT cisternography for an epidermoid, and was a rare case due to the coexistence of trigeminal neuralgia and ipsilateral hemifacial spasm. It should be kept in mind in view of this case that a brain tumor may be one of the possible causes of trigeminal neuralgia and/or hemifacial spasm.

References

1) Cook BR, Jannetta PJ: Tic convulsif: Results in 11 cases treated with microvascular decompression of the fifth and seventh cranial nerves. J Neurosurg **61**: 949-951, 1984.
2) Cushing H: The major trigeminal neuralgia and their surgical treatment based on experiences with 332 gasserian operations. First paper. The varieties of facial neuralgia. Am J Med Sci **160**: 157-184, 1920.
3) Gardner WJ: Trigeminal neuralgia. Clin Neurosurg **15**: 1-56, 1968.
4) Imamura Y, Ninchoji T, Nakajima S, et al: Epidermoid tumor in the fourth ventricle, with particular reference to metrizamide CT cisternography findings. Surg Neurol **18**: 444-447, 1982.
5) Maurice-Williams RS: Tic convulsif: The association of trigeminal neuralgia and hemifacial spasm. Postgraduate Med J **49**: 742-745, 1973.

6) Niizuma H, Ikeda S, Ohyama H: Trigeminal neuralgia and hemifacial spasm caused by the compression of tortuous vertebro-basilar system. A case report. Neurol Surg **9**: 1167-1170, 1981.
7) Revilla AG: Tic douloureux and its relationship to tumors of the posterior fossa. Analysis of twenty-four cases. J Neurosurg **4**: 233-239, 1947.
8) Sekihara Y, Takeda N, Tanaka R, et al: Two cases of epidermoid in the fourth ventricle with characteristic findings of metrizamide CT cisternography. Prog Comp Tomo **5**: 721-725, 1983.
9) Wakabayashi T, Tamaki N, Satoh H, et al: Epidermoid tumor presenting as painful tic convulsif. Surg Neurol **19**: 244-246, 1983.

和文抄録

三叉神経痛と同側の顔面痙攣をきたした Epidermoid の1例

神戸市立中央市民病院　脳神経外科
大塚　信一，中津　正二，松本　茂男，佐藤　慎一
本崎　孝彦，伴　　貞彦，山本　豊城
神戸市立中央市民病院　臨床病理科
白　根　博　文

Painful tic convulsif をきたした epidermoid の1例を報告した．35歳男性で，右三叉神経痛と同側の顔面半側痙攣を症状とし，術前 CT および metrizamide CT cisternography で，epidermoid と診断され，手術で epidermoid であることが確認され，術後症状は完全に消失した．本症例は metrizamide CT cisternography が epidermoid に特徴的な所見を示し，術前診断として非常に有用であった．三叉神経痛，顔面痙攣の症例の中に脳腫瘍によるものがあることを念頭におく必要があると考える．